

Case Report

Mucoepidermoid carcinoma of the parotid gland mimicking a benign cystic lesion

Gokul Sridharan ^{a,*}, Kaveri Hallikeri ^b

^aDepartment of Oral and Maxillofacial Pathology, Rajah Muthiah Dental College and Hospital, Annamalai University, Annamalai Nagar, Chidambaram, Tamilnadu

^bDepartment of Oral & Maxillofacial Pathology, S.D.M College of Dental Sciences and Hospital, Sattur, Dharwad

Abstract. Mucoepidermoid carcinoma is the most common malignant salivary gland neoplasm. It affects both major and minor salivary glands and when major salivary glands are affected, the parotid gland is the most common. The incidence has been reported to range from 4-12% of all the parotid tumors. Mucoepidermoid carcinoma of parotid gland is usually of low grade type with a marked cystic component causing considerable diagnostic difficulties. Hence cystic lesions of parotid gland should not be merely dismissed as benign entities and mucoepidermoid carcinoma should be always included in differential diagnosis.

In this presentation, we report a case with initial diagnosis of benign cystic lesion based on FNAC and MRI findings which was later diagnosed histologically as low grade mucoepidermoid carcinoma of the parotid gland.

Key words: Mucoepidermoid carcinoma, mucocele, cystic lesions, parotid gland

1. Introduction

The cysts of the parotid gland are rare lesions. The symptoms include a slowly growing painless mass without fixation to the overlying skin or involvement of the facial nerve. Such cystic lesions especially mucocele must be distinguished from salivary gland neoplasms with similar clinical presentation and association with a cystic component. Making a right preoperative diagnosis can be extremely difficult but is of paramount importance as they dictate the treatment modality. However the final diagnosis is established histopathologically and satisfactory therapy remains the superficial parotidectomy. This article presents a case that was presumed to be a mucocele based on clinical, radiographic and FNAC findings, but was histopathologically diagnosed as a cystic/low grade mucoepidermoid carcinoma.

2. Case report

A 64 year old male patient presented with a painless, well defined solitary swelling of right parotid region of two and half years duration, measuring 5x4cm in size, slightly tender and not fixed to underlying structures (Fig 1).



Fig. 1. Well defined solitary swelling of the right parotid region (arrow).

There was no history of discharge, xerostomia and the lesion was neither compressible nor reducible. Facial nerve function was intact. Patient's medical history revealed hypertension and coronary artery disease and he was on medications for the same diseases. A clinical differential diagnosis of mucocele and benign salivary gland tumors like pleomorphic adenoma and Warthin's tumor was considered.

*Correspondence: Dr. Gokul Sridharan
Department of Oral and Maxillofacial Pathology
Rajah Muthiah Dental College and Hospital
Annamalai University,
Annamalai Nagar,
Chidambaram, Tamilnadu
Tel.: 09940195372;
E-mail: drgokuls@gmail.com
Received : 08. 07. 2010
Accepted : 18.08.2010

MRI findings showed a well defined cystic space occupying lesion in superficial portion of parotid gland measuring 4x3x3cm. The lesion was hypodense on T1- weighted image and hyperdense on T2- weighted image (Fig 2).

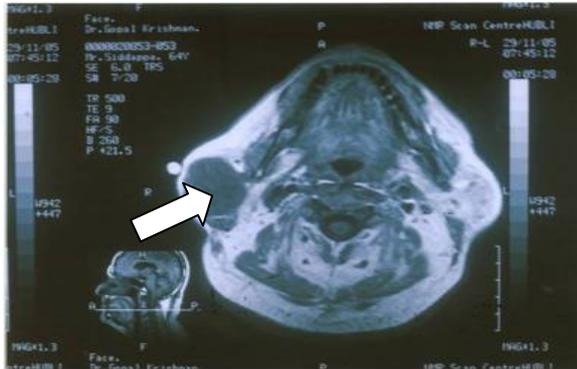


Fig. 2. Well defined cystic space occupying lesion in superficial portion of parotid gland. The lesion was hypodense on T1- weighted image (arrow).

Fine needle aspiration cytology (FNAC) performed prior to surgery revealed mucinous material filled with mucinophages. The MRI and FNAC findings were suggestive of benign cystic lesion. Based on this diagnosis superficial parotidectomy was done. The surgical margins were clean and the facial nerve was intact without any tumor involvement.

On macroscopic examination the lesion measured around 7x5cm, brownish in color, soft in consistency with outer granular surface and regular borders. The cut surface showed a well defined single large cystic cavity with no obvious proliferative growth (Fig 3), filled with a greenish mucous substance was present with a jelly like consistency.

Pathologic evaluation of the resected parotid mass showed multiple cysts of varying size within the fibrous connective tissue, squamous and intermediate cells (Fig 4).



Fig. 3. cut surface shows a single large cystic lumen.

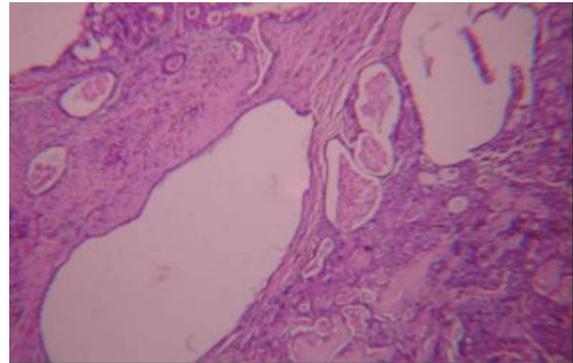


Fig. 4. (H&E 10x): Multiple cystic spaces of varying size with mucus and epidermoid cells.

Large cystic lumens lined by squamous and mucous cells were noticed (Fig 5). Squamous atypical cells were present among these cystic spaces. There was no evidence of mitotic figures or necrosis. The final diagnosis was thus considered as cystic, low grade mucoepidermoid carcinoma of parotid gland.

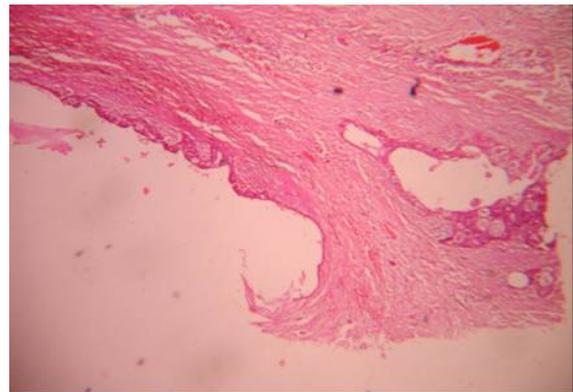


Fig. 5. (H&E 10x): Large cystic lumen lined by epidermoid and mucus cells.

The patient was then subjected to radiotherapy and was followed up. Follow up until six months did not reveal any recurrence and the healing was uneventful (Fig 6).



Fig. 6. Post operative follow up.

3. Discussion

A mass in the salivary gland region presents a diagnostic challenge with regard to its site of origin, nature of the lesion, behaviour and tissue specific diagnosis. A variety of neoplastic and non-neoplastic lesions of salivary glands present with a prominent cystic architecture (1). FNAC has a recognized role in the evaluation of all salivary gland swellings. The use of FNAC in diagnosing cystic lesions of the parotid gland is a common and appropriate practice. However use of FNAC alone for accurate diagnosis may be difficult (2). FNAC of salivary gland lesions may yield watery or mucoid material frequently of low cellularity. The cellularity may be scant or absent making cytological diagnosis difficult and at times impossible (1).

Several authors have reviewed the FNAC findings of salivary gland lesions, the sensitivity and specificity of which have been widely reported in the literature. According to Edwards PC et al (2005) (3) the accuracy of diagnosing salivary gland lesions with the help of FNAC is more than 70%. Stewart CJ et al (2000) (4) opined that the overall sensitivity, specificity and accuracy of FNAC diagnosis were 92%, 100% and 98% respectively. In another study by Jayaram G et al (1994) (5) in FNAC diagnosis of 241 salivary gland lesions the accuracy rate was 91%. The sensitivity and specificity rate for malignant tumors were 87.8% and 98%. For benign tumors 100% sensitivity was observed.

Despite the high percentage of accuracy obtained with the use of FNAC, the possibility of false positive or false negative results cannot be ruled out. It is emphasized that the growth of the tumor may play a significant role in the obstruction of salivary gland duct lead to cyst formation which may further complicate diagnosis. Also the presence of cystic structures within the benign and malignant tumors may create diagnostic difficulties.

Salivary gland tumors account for less than 5% of head and neck neoplasms. Among these mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumor. About 45% of MEC occur in the parotid gland and appear around 5th decade of life (6). Histologically MEC is divided into 3 grades based on presence of cystic structures, cellular atypia and cell type into low grade, intermediate grade and high grade tumors. Low grade tumors arise more commonly in minor salivary glands while high grade tumors are seen in major salivary glands (7). Prominent cystic structures are the hallmark of low grade MEC and several

large cysts may form a significant portion of tumor, sometimes occupying more than 50% of histological sections (2). The presence of these cystic structures may lead to underdiagnosis which was evident in the present case. Hence a thorough evaluation of the cystic lesions of the parotid gland is to be done with consideration of malignant lesions in the differential diagnosis. The treatment usually consists of surgery followed by radiotherapy to minimize post-operative recurrence. Other possible conditions where radiotherapy may be required include close surgical margins and perineural invasion.

MEC have been reported to show highest false negativity with FNAC among all the salivary gland neoplasms. This could be attributed to the presence of cystic architecture and cellular dilution due to cyst fluid causing failure to detect the epithelial cells (3). In the present case, a preliminary diagnosis based on clinical, radiographic and FNAC findings was given as benign cystic lesion probably mucocele. Thus the cystic lesions of parotid gland though showing benign behaviour, a differential diagnosis of low grade MEC should be considered. The differential diagnosis for cystic lesions of the parotid gland includes benign conditions like mucocele, lymphoepithelial cysts, Warthin's tumor, lymphangioma, dermoid cysts, parotid cysts and malignant conditions like low grade MEC. Knowledge of cytological features of MEC and its differential diagnosis is mandatory for accurate diagnosis. Smears in MEC contain cohesive sheets of tumor cells with abundant cytoplasm showing squamous differentiation as well as cells containing intracytoplasmic mucin vacuoles (8). Identification of 3 cell types: intermediate, mucus producing and squamous cells in the smear is the most important diagnostic criteria. Presence of overlapping epithelial groups is also an important feature (9). Cytological differential diagnosis for MEC includes non-neoplastic conditions like mucus retention cysts, chronic sialadenitis, lymphoepithelial cysts and neoplastic conditions like Warthin's tumor, pleomorphic adenoma, cystadenoma and cystadenocarcinoma (1).

The presence of thick mucinous material in the background associated to a scantily cellular smear will lead to underdiagnosis of low grade MEC as a benign cystic lesion. In such cases, the presence of extracellular mucin in combination with intermingling intermediate cells and mucus producing cells should be identified to make a correct diagnosis (9). The presence of chronic inflammation in an aspirate may be suggestive of sialadenitis though low grade MEC may show

chronic inflammatory cell reaction (2). Stringy mucin seen in MEC may be helpful in differentiating it from the mucin seen in pleomorphic adenoma. The diagnosis may be difficult in the presence of mucinous metaplasia. Also the myxoid ground substance can be mistaken for epithelial mucin leading to misdiagnosis of pleomorphic adenoma as malignant lesion. The presence of chondromyxoid material can serve as a useful clue for accurate diagnosis (9). Other differential diagnosis includes mucinous adenocarcinoma and mucinous cystadenocarcinoma.

Mucinous cystadenocarcinoma must be always cystic while the size and prominence of cysts present in low grade MEC varies. Mucinous adenocarcinoma is not cystic but gelatinous. Nuclei are bland in both mucinous cystadenocarcinoma and low grade mucoepidermoid carcinoma but are atypical in mucinous adenocarcinoma. There is no squamous differentiation in either mucinous cystadenocarcinoma or mucinous adenocarcinoma, but this latter is subtle in low grade mucoepidermoid carcinomas. Mucinous cystadenocarcinoma should be considered a potential candidate in the differential diagnosis of mucinous lesions that can occur in the salivary gland (10). The large number of differential diagnosis for parotid gland swellings and the fact that the clinical and radiographic findings are often indefinite make a conclusive pre-operative diagnosis often difficult. CT and MRI can describe the location of the mass and its relation to the surrounding structures but may be insufficient for a final pre-operative diagnosis. FNAC may be helpful but the possibilities of false negative and false positive results exist. As evident from the present case, low grade MEC of the parotid region mimics a benign cystic lesion based on MRI and FNAC findings and hence

histopathological confirmation is necessary for final diagnosis. Also cystic lesions of parotid gland should be regarded as neoplasm until proven otherwise. Mucoepidermoid carcinoma should be considered in the differential diagnosis of any mucocele like mass of the parotid gland.

References

1. Layfield LJ, Gopez EV. Cystic lesions of the salivary glands: cytologic features in fine-needle aspiration biopsies. *Diagn Cytopathol* 2002; 27: 197-204.
2. Awwad RJ, Hsu J. Mucoepidermoid carcinoma of the parotid gland: an unusual presentation. *Am J Otolaryngol* 2006; 27: 344-345.
3. Edwards PC, Wasserman P. Evaluation of cystic salivary gland lesions by fine needle aspiration: an analysis of 21 cases. *Acta Cytol* 2005; 49: 489-494.
4. Stewart CJ, MacKenzie K, McGarry GW, et al. Fine-needle aspiration cytology of salivary gland: a review of 341 cases. *Diagn Cytopathol* 2000; 22: 139-146.
5. Jayaram G, Verma AK, Sood N, et al. Fine needle aspiration cytology of salivary gland lesions. *J Oral Pathol Med* 1994; 23: 256-261.
6. Clarós P, Dominte G, Clarós A, et al. Parotid gland mucoepidermoid carcinoma in a 4-year-old child. *Int J Pediatr Otorhinolaryngol* 2002; 63: 67-72.
7. Kokemueller H, Brueggemann N, Swennen G, et al. Mucoepidermoid carcinoma of the salivary glands—clinical review of 42 cases. *Oral Oncol* 2005; 41: 3-10.
8. Das KA, Petkar MA, Nadra M, et al. Role of Fine Needle Aspiration Cytology in the Diagnosis of Swellings in the Salivary Gland Regions: A Study of 712 Cases. *Med Princ Pract* 2004; 13: 95-106.
9. Rupani BA, Kavishwar VS, Achinmane V, et al. Fine needle aspiration cytology of low-grade Mucoepidermoid carcinoma of the parotid gland: A diagnostic challenge *J Cytol* July 2008; 25: 115-116.
10. Schneller J, Solomon M, Webber CA, et al. Mucinous cystadenocarcinoma of the parotid gland: report of a case with fine needle aspiration findings and histologic correlation. *Acta Cytol* 2001; 45:605-609.